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Reactive Attachment Disorder in Maltreated Infants and Young Children in Foster Care

Dr Molly Bruce*

NHS Lothian

Dr David Young

University of Strathclyde

Dr Susan Turnbull

Dr Maki Rooksby

Mr Guy Chadwick

Dr Catriona Oates

Ms Rebecca Nelson

Dr Genevieve Young-Southward

Dr Caroline Haig and

Prof. Helen Minnis

Institute of health and wellbeing, University of Glasgow

*Address for Correspondence: Clinical Psychology, Western General Hospital, Edinburgh, EH4 2XU. E-mail: molly.bruce@nhslothian.scot.nhs.uk

Abstract

Background: Reactive attachment disorder (RAD) has been described as one of the least researched and most poorly understood psychiatric disorders. Despite this, given what is known about maltreatment and attachment, it is likely that RAD has profound consequences for child development. Very little is known about the prevalence and stability of RAD symptoms over time. Until recently it has been difficult to investigate the presence of RAD due to limited tools for informing a diagnosis. This study utilised an observational tool, the Rating of Inhibited Attachment Behavior (RInAB), which has recently been developed by experts in the field.

Method: a short-term prospective longitudinal study explored RAD symptoms in maltreated infants in Scotland (n=100, age range= 12-62 months) over 12 months. Relationships between RAD symptoms and mental health and cognitive functioning were also considered. The study utilised the RInAB (Corval, Baptista, Fachada, Beiramar, & Soares, in press) alongside The Disturbances of Attachment Interview (Smyke & Zeanah, 1999). Children were recruited as part of the Best Services Trial (*BeST²*) study, in which all infants who came in to the care of the local authority in Glasgow due to child protection concerns were invited to participate. **Results:**

Prevalence of RAD was found to be 5.0% (n=5, 95% CI [0.7 – 9.3]) at T1, when children are first placed in to foster care. At T2, following at least one year of improved care conditions, 2.1% (n=2, 95% CI [below 0 – 4.7]) met diagnostic criteria for RAD. RAD was associated with some mental health and cognitive difficulties. Observed symptoms and carer-reported symptoms were uncorrelated. While levels of carer-reported RAD symptoms decreased significantly over time, observed symptoms did not. **Conclusions:** Our findings suggest that RAD resolved in a small majority of the few cases that initially met the diagnostic criteria. Further exploration in larger samples would be invaluable.

Keywords: *Reactive attachment disorder, child maltreatment, inhibited attachment, foster care*

Reactive Attachment Disorder in Maltreated Infants and Young Children in Foster Care

Attachment is a fundamental process across species whereby, for protection and survival, an infant seeks to be close to its caregiver when distressed (Bowlby, 1969). In humans, the formation of a secure attachment relationship allows for positive social development and emotional regulation, which protects against mental health problems (Prior & Glaser, 2006). In reactive attachment disorder (RAD), there appears to be no activation of the attachment system. Children with RAD do not appear to demonstrate important attachment behaviours such as seeking or accepting comfort, or signalling distress when frightened (Zeanah & Gleason, 2015). It is proposed that RAD exclusively occurs in the context of maltreatment, where the infant's attachment behaviours have been consistently neglected from a young age (The Diagnostic and Statistical Manual of Mental Disorders, 5th ed.; DSM-5; American Psychiatric Association [APA], 2013). Children with RAD may be socially and emotionally withdrawn in a wide range of situations, which is likely to limit their ability to make use of love or care from others and reduce opportunities for learning. Therefore, RAD may well have a considerable detrimental effect on child development (Prior & Glaser, 2006).

RAD was first defined in 1980 and the diagnostic criteria have been revised several times since (Zeanah & Gleason, 2010). It has been described as “one of the least researched and most poorly understood disorders listed in the DSM” (Chaffin, et al., 2006). There were previously two forms of the disorder as defined by the DSM-IV and DSM-IV-TR (APA, 1994, 2000) these being ‘inhibited reactive attachment disorder (I-RAD)’ and ‘disinhibited reactive attachment disorder (D-RAD)’. The ICD-10 (WHO, 2010) divided the subtypes into two distinct disorders and more recently the DSM-5 (APA, 2013) has similarly updated its classifications. The previously termed inhibited form is now defined as ‘reactive attachment disorder’ (RAD) and the previously termed disinhibited form is now classified as ‘Disinhibited Social Engagement Disorder’ and is no longer considered a disorder of attachment (Zeanah & Gleason, 2015). The

DSM-5 defines RAD as a consistent pattern of inhibited, emotionally withdrawn behaviour towards adult caregivers, and persistent social and emotional disturbance, in the context of extreme patterns of insufficient care. The DSM-5 goes on to note that for a diagnosis of RAD, the disturbance must be evident between the ages of nine months and five years. The ICD-10 details that children with RAD may exhibit misery, huddling, clinginess, an inappropriate lack of response, or aggression. Zeanah and Gleason (2015) add that features of RAD include unexplained fearfulness or irritability.

Researchers are reasonably confident about the prevalence of DSED (Boris et al., 2004). The prevalence of RAD, however, is unknown but appears to be a rarer disorder (Corval et al., in press; Gleason et al., 2011). The DSM-5 states that less than 10% of children who have been severely neglected develop RAD and it is considered to be most common in children with an experience of institutionalisation (Corval et al., in press). Only a relatively small number of studies have investigated the prevalence of RAD distinctly and of those that have, the findings vary widely across studies. Furthermore, it is difficult to make comparisons across studies given the considerable differences in care settings, historical care settings, role of informants, measures used to inform RAD, and use of diagnosis versus symptomology of RAD. In foster care samples, the prevalence of RAD has been found to vary between 3% and 35% (Jonkman et al., 2014; Zeanah et al., 2004). Few studies have explored prevalence over time. In conducting the Bucharest Early Intervention Project with previously institutionalised Romanian children, Gleason et al. (2011) explored RAD over time and found that the number of children meeting diagnostic criteria varied at each time point (4.6% at baseline, 3.3% at 30 months, 1.6% at 42 months and 4.1% at 54 months). Zimmerman (2016) investigated RAD in foster children over one year and found a prevalence of 5.5% (n=3) shortly after placement and 1.9% (n=1) 12 months after placement.

Some researchers argue that further clarity around the definition of RAD is needed. Zeanah and Gleason (2010) propose that symptoms of RAD are signs of current maltreatment

rather than a persistent disorder. Zeanah, Mammen and Lieberman (1993) assert that the frozen watchfulness associated with RAD is, in fact, a response when confronted by an abusive caregiver rather than an expressed sign of attachment disorder. If RAD were simply a 'state' associated with current maltreatment, it may be expected to disappear once a child is placed in a stable, nurturing foster family. Indeed Jonkman et al. (2014) found that if children experienced an improvement in caregiving conditions (being placed in foster care) RAD was less persistent than DSED. Jonkman et al. (2014) went on to report that there was a negative association between RAD symptoms and time in foster care placement, and that RAD ultimately disappeared. Other studies however have found that although prevalence of RAD decreases, RAD persists for some children after one year in a foster care placement (Zimmermann, 2016). Therefore, it is unclear whether RAD is a *state* associated with current maltreatment or a *disorder* that is pervasive across time and contexts.

Until recently, it has been difficult to investigate the presence of RAD due to limited measures for informing a diagnosis specific to RAD and because symptoms are subtle, there is no consensus about whether carer report, observation or both are most crucial. The Rating of Inhibited Attachment Behavior (RInAB) (Corval et al., in press), an observational tool for the assessment of RAD has now been developed by a group of experts in the field.

There is limited research exploring the mental health of children with RAD, however behaviours indicative of attachment disorders have been shown to be distinct from conduct problems, emotional problems and hyperactivity (Minnis et al., 2007). Yet, given the link between early childhood psychopathology and difficulties in the parent-child relationship (Skovgaard et al., 2007), it is likely that children with symptoms of RAD have a higher likelihood of experiencing mental health difficulties. Millward, Kennedy, Towlson, and Minnis (2006) found a significant association between measures of attachment disorders and other mental health symptoms ($r = .84$), however, this study was not specifically exploring RAD. Moran, McDonald, Jackson, Turnbull, and Minnis (2017) explored RAD independently in a

youth justice population (12-17 years) and found a strong association between RAD and other mental health symptoms with a large effect size ($r_s = .60$).

Research investigating RAD specifically has shown that it is associated with: depressive symptoms (Gleason et al., 2011); social difficulties (Elovainio, Raaska, Sinkkonen, Mäkipää, & Lapinleimu, 2015; Gleason et al., 2011); and poorer psychological wellbeing (Elovainio et al., 2015; Minnis et al., 2007; Moran et al., 2017). There are however conflicting findings in relation to RAD and its relationship with emotional difficulties, internalising difficulties and externalising difficulties. Elovainio et al. (2015) and Gleason et al. (2011) found an association between RAD and emotional problems whereas Moran et al. (2017) did not. While some studies have found associations between RAD and internalising difficulties (Elovainio et al., 2015; McGoron et al., 2012), others have not (Jonkman et al., 2014; Lehmann, Breivik, Heiervang, Havik, & Havik, 2016). It should be noted that studies finding no associations often reported low levels of RAD symptoms in their samples (for example Lehmann et al., 2016 & Jonkman et al., 2014) and had smaller sample sizes; which may increase the risk of a type II error, particularly when exploring RAD as a categorical diagnosis. Elovainio et al. (2015) reported an association between RAD and externalising difficulties, however McGoron et al. (2012) did not find such relationship. Differing findings in relation to RAD and mental health may also be due to the widely differing age ranges of samples, warranting further longitudinal research.

Furthermore, research has found that RAD is associated with lower cognitive ability in institutionalised children (Smyke, et al., 2012). Other studies have found that RAD and DSED collectively (Pritchett, Pritchett, Marshall, Davidson, & Minnis, 2013) and RAD independently (Gleason et al., 2011) are associated with below average cognitive functioning. Studies combining both types of RAD have demonstrated associated language difficulties (Minnis et al., 2009; Sadiq et al., 2012). It would be useful to further consider the relationship between cognitive functioning, verbal comprehension and RAD specifically in a non-institutionalised

sample.

In summary, there is insufficient evidence regarding the prevalence and stability of RAD symptoms, particularly in non-institutionalised samples of maltreated children. Furthermore, very little is known about the relationships between RAD, other mental health problems and cognitive functioning. This study is an attempt to address some gaps in the scientific literature and investigate RAD in maltreated infants over a one-year time period. The primary aim was to establish the prevalence and stability of RAD symptoms in a maltreated sample, comparing symptoms shortly after placement in foster care (Time 1) to the level of symptoms exhibited after one year in foster care (Time 2). A secondary aim was to make a preliminary exploration of the relationships between symptoms of RAD and mental health difficulties and cognitive functioning.

Research Questions

1. What is the prevalence of RAD in young children and infants in foster care and how do symptoms change over time? Our hypothesis here is that the level of RAD symptoms will reduce over time but clinical levels of symptoms will remain, over one year, for some
2. How do RAD symptoms correlate with mental health difficulties and cognitive functioning? Our hypothesis here is that symptoms of RAD will be significantly associated with other mental health difficulties and lower cognitive functioning.
3. Do observation and carer report of RAD symptoms show concordance? Our hypothesis here is that observed symptoms will differ from those reported by carers because RAD symptoms are subtle and easy to miss.

Method

Participants

In this prospective longitudinal study, the baseline sample consisted of 100 children aged between 12 and 62 months. All participants had been accommodated in to the care of local authority Social Work in the Scottish city of Glasgow and recruited for The Best Services Trial (BeST[?]); an ongoing randomised controlled trial investigating an infant mental health intervention (clinical trials registration number NCT01485510). Careful review of the quality of foster placements took place and children were only returned to birth families when it was considered that they were sufficiently reformed to provide nurturing care. Demographic information relating to the sample is provided in Table 1.

Inclusion and exclusion criteria. All parents or guardians of a child aged between approximately 6 - 60 months who was placed in local authority care for a period due to child protection concerns were invited to take part in the BeST[?] Trial (Pritchett et al., 2013); from which data are being examined here. Children were excluded from the trial if they had a profound learning disability (as some assessment measures would not be appropriate) or their primary caregiver was unavailable to take part (such as long-term imprisonment, death, or being uncontactable by services or the research team for 3 months or more).

Additional exclusion criteria for the current study: Children under 12 months old were excluded due to selective attachment behaviours still potentially being developed up until this age (Bowlby, 1969), therefore attachment measures may not be appropriate for infants under 12 months. One child was excluded because of Autism Spectrum Disorder since the observational measure utilised advises that it should only be used to assess children with no sensory, neurological or genetic disorders.

Recruitment Procedures

Recruitment (for the BeST[?] Trial) took place between December 2011 and April 2013, and each eligible child who entered care due to child protection concerns during this period was considered for participation. Birth parents and foster carers of eligible children were approached

to discuss the BeST² Trial by a recruitment officer (a specially trained social worker), who provided an information leaflet and a video explaining the study. Informed consent was gained from those families willing to participate. It was made clear to the carers and birth parents of the eligible participants that participation was entirely voluntary and would not affect any aspect of their care or management. The West of Scotland NHS Research Ethics Committee, 5, approved the study.

A power calculation was carried out for the hypothesis that RAD scores would change over time. A previous study (Millward, et al., 2006) found a correlation of 0.84 (equating to an effect size of around 0.7) between RAD and SDQ scores. A sample size calculation performed using G*Power 3.1 (Faul, Erdfelder, Lang, & Buchner, 2007), with power of 0.8 and significance level of 0.05, concluded that a sample size of 76 was sufficient to detect a moderately high but conservative effect size of 0.8.

At time of data collection, the BeST² Trial had data for N=101 children with a recruitment rate of 58% of eligible families at baseline (T1) and a retention rate of around 79% of eligible families at 1 year follow-up (T2). A researcher in the team attempted to establish reason for drop-out or non-follow up at T2. Most often dropout was due to birth parent(s) being un-contactable at this time point (10%) or withdrawing consent (4.1%), or participants being excluded following baseline assessment, for example if courts failed to establish grounds for the child being in local authority care (3.6%). It should be noted that independent samples Mann-Whitney U tests demonstrated no significant difference between those who continued to participate in the study and those who dropped out following T1 in observed RAD symptoms at T1 ($p=0.61$), carer-reported RAD symptoms at T1 ($p=0.52$), age ($p=0.56$) and gender ($p=0.94$). See Figure 1 for a visual representation of the participants included in the data set.

Baseline assessments were administered approximately three months after the child was placed into care (T1). This time period allowed the child to begin to settle in to the placement and for the child and carer to become familiar with each other. Follow up assessments were

completed approximately one year after their T1 assessment (T2). Assessments were completed with participating children and their primary caregiver. At baseline this was the same foster carer, at follow up in most instances (73.1%) this was still their foster carer but in some cases it was a new foster carer, birth parent, adoptive parent, or kinship carer (as indicated in table 1). If the carer at T2 was different to the T1 carer, the child had to be placed with them for at least four weeks before the T2 assessment was completed however for those included in this study, it had usually been significantly longer.

Measures

The Rating of Inhibited Attachment Behavior Scale (RInAB; Corval et al., in press) (Appendix 1.1) was administered at T1 and T2 using video footage. The RInAB is an observational measure of RAD for preschool-aged children, which has been developed based on scientific literature, DSM-5 criteria, and repeated observations of interactions of child-caregiver dyads (Corval et al., in press). The scale contains 17 items rated between 0 (not at all characteristic) and 4 (very characteristic), grouped into three sub-scales: Attachment behavior, Exploratory behaviour and Socioemotional behaviour. The authors developed the RInAB tool for use with three to six year olds. In the absence of any other observational measure for reactive attachment disorder, this study used the RInAB with children younger than the authors of the tool advise. The observational tool seemed to be equally as useful with the younger age group given attachment behaviours have generally developed by the age range included in the study (12-35 months). Although attachment behaviours may vary depending on developmental stage, no adaptations to the coding were required. Another paper in this special issue concerns the validity of the RInAB (Corval et al., in press). The tool can be used with The Strange Situation Procedure (Ainsworth & Bell, 1970) and other situations that activate the attachment system. For the current study, SSP video recordings were used to inform RInAB rating when available; formal SSP scoring did not take place as this was not the focus of this study. 55% of participants

had an SSP available at T1, T2 or both. The remaining 45% of participants had laboratory video footage available that included separations and reunions with caregivers. No significant differences in RInAB scores at T1 or T2 were found between those who did and did not have an SSP administered (T1: median= 0.73 (0.41, 1.38) versus median= .54 (0.29, 0.88), $U(99)=355.5$, $p=.28$, $r=.10$) (T2: median= 0.46 (0.23, 0.69) versus median= 0.33 (0.25, 0.66), $U(75)=582.5$, $p=.76$, $r=.03$).

The Disturbances of Attachment Interview (DAI) (Smyke & Zeanah, 1999) was administered at T1 and T2. The DAI is a semi-structured interview for attachment disorder behaviours, designed to be administered by clinicians to caregivers. This study focused on the emotionally withdrawn/inhibited subscale: these five items explore how well the child differentiates among adults, whether the child shows a clear preference for a particular caregiver, seeks comfort from a preferred caregiver, responds to comforting when offered, the degree to which the child responds reciprocally in social interactions and whether the child shows developmentally appropriate levels of emotional regulation. This scale produces scores of 0 to 10, with higher scores reflecting increasing signs of RAD. Smyke, Dumitrescu and Zeanah (2002) reported that the DAI scales demonstrate strong internal validity for RAD (Cronbach's α 0.80) and that inter-rater reliability was excellent ($\kappa=0.88$). DAI's were available for 94% ($n=94$) of participants at T1 and 96% ($n=73$) at T2. Missing DAI data was due to carers being unavailable on numerous occasions.

The Strengths and difficulties questionnaire (SDQ) (Goodman, 1997) was administered at T2. The SDQ is a brief behavioural screening questionnaire for 2-17 year olds. It includes 25 items involving psychological attributes, divided between five scales: emotional symptoms, conduct problems, hyperactivity, peer relationship problems and prosocial behaviour. Subscales can be used individually and the first four subscales are summed to form a Total Difficulties

Score. The SDQ has been well validated across a wide age range by various studies (Goodman, 2001).

The Wechsler Pre-school and Primary Scale of Intelligence (WPPSI IV) (Wechsler, 1967), a cognitive assessment for use with children between the ages of 30 and 91 months old, was administered at T2. The assessment generated Full-scale IQ, Performance IQ and Verbal IQ scores. One child was younger than the minimum age for this assessment at T2 and therefore was not assessed.

Although all children received a RInAB at both T1 and T2 (n=100 at T1; n=76 at T2), there were some missing data for the other measures – please see Figure 1 for detail.

Clinical Diagnosis

As in Gleason et al. (2011) the research diagnostic criteria for RAD were applied to create a categorical indicator of carer-reported RAD following DSM 5 criteria. Using the DAI, for a child to meet DSM 5 criteria, the three DAI items most central to RAD (items 1, 2a, 3) (APA, 2013) had to be met. Similarly, using the RInAB, the authors state that to meet DSM 5 criteria, two items must be endorsed in the Attachment Behaviour Subscale (items 1 & 2) in addition to any two items in the Socioemotional Behavioural Subscale. Where observational criteria (informed by the RInAB) and carer-report criteria (informed by the DAI) were met, cases were discussed with a supervisor of the project (Child Psychiatrist specialising in RAD) and a multi-informant, clinical diagnosis was given. A borderline diagnosis of RAD was given if there was substantial disagreement in observed and carer-reported symptoms (i.e. where it was clear that only carer-report criteria or only observational criteria were met) or if from the information available, some elements of RAD remained unclear.

Research Procedures

Baseline assessments were administered approximately three months after the child was placed into foster care (T1). This time period allowed the child to begin to settle in to the placement and for the child and carer to become familiar with each other. Follow up assessments were completed approximately one year after their T1 assessment (T2). Assessments were completed with participating children and their primary caregiver. In most instances (73.1%), the carer at T1 remained the same at T2 but in some cases it was a new foster carer, birth parent, adoptive parent, or kinship carer (as indicated in table 1). If the carer at T2 was different to the T1 carer, the child had to be placed with them for at least four weeks before the T2 assessment was completed.

Each video clip, of which RInAB ratings were based on, was approximately 30-50 minutes in duration. Footage included approximately 20 minutes of the infant playing with their caregiver using a set of age-appropriate toys and around 20 minutes of them having a lunch together; both toys and lunch were standard and provided by the research team. Separations and reunions as detailed below were also captured. Attachment stress was introduced by the entry of a stranger into the room and a separation, if a Strange Situation Procedure was also available (n=65), there were a series of separations and reunions. Where an SSP was available, the standard method was used for participants aged 12-18 months (Ainsworth, 1970). For participants older than 18 months, the preschool method of the SSP was administered (Cassidy & Marvin, 1992) unless there was thought to be developmental delay, in which case the infant version was used. The preschool SSP involved longer separations and no stranger. Regardless of whether or not an SSP was administered, each participant was separated from their caregiver for approximately one hour whilst they completed a cognitive assessment with a researcher (a 'stranger'), they were reunited afterwards. It was felt that these separations and reunions were an ample opportunity to observe attachment behaviours. On two occasions, the carer was invited to join the participant for their cognitive assessment due to significant distress on separation. The RInAB scale was administered for each participant using the recordings of child-carer dyads.

Attempts were made to keep raters blind to whether the footage being observed was at T1 or T2, but because of the rarity of children with RAD and the sometimes striking nature of their behaviours, this was not always possible. Trained researchers provided inter-rater by scoring 20% of the sample across T1 and T2 (n=35), including a range of cases that did and did not meet observational criteria for RAD. Training for the RInAB was provided by the main author of the tool as was some inter-rater reliability where individual scoring was compared and discussed. Supervision of rating was provided by HM. In relation to the administration of the observational measure of RAD (RInAB), a good level of inter-rater reliability was found (95%; Kappa=0.9) and any discrepancies were resolved through discussion.

Statistical Analysis

Given that this is the first study in the UK to consider the prevalence and stability of RAD and associated difficulties, it should be considered as an exploratory study that will inform future research.

Distributions of continuous and discrete variables were explored graphically and using Kolmogorov-Smirnov tests. Assumptions of normality were not met and due to this and the discrete nature of the outcomes, non-parametric statistical tests were used. Within-group comparisons between symptoms at T1 and T2 were carried out using the paired Wilcoxon Signed Ranks test. Mann-Whitney Tests were used to make independent group comparisons between those whose symptoms improved and those whose did not. Effect sizes, r , were calculated using the Rosenthal (Rosenthal, 1994) formula for non-parametric data where 0.1, 0.3 and 0.5 indicate small, medium and large effect sizes respectively (Cohen, 1988).

Spearman's correlations were used to assess multicollinearity and therefore reduce the number of explanatory variables used in the modelling and enhance power. Regression analyses were considered but given the non-parametric nature of the data and the sample size, such tests

were ruled out. Because the age-range of the sample was large, we considered whether age was associated with all relevant variables (RInAB, DAI, SDQ) to ensure the associations were not confounded by age. All statistical procedures were performed using SPSS version 22.

Results

Research Question 1: Prevalence and Stability of RAD

In order to examine prevalence at T1 and prevalence T2, we regarded the T1 and T2 samples as two independent cross-sectional samples. This was for two reasons: 1. there was a higher RInAb score in those who dropped (mean 0.75 out by T2 compared those who did not and 2. one of the five children who met diagnostic criteria at T1 was no longer in the study by T2. The median score on both the RInAB and DAI was higher in the 100 children examined at T1 and was lower in the 76 children examined at T2 (see Table 2).

In order to investigate stability of RAD symptoms, only those with data at both time points were included. The difference was not significant for observed symptoms (RInAB) (0.56 T1 vs 0.46 Ts; $Z = -1.67$; $p = .01$, $r = 0.19$) and although there was a significant reduction in carer-reported symptoms (DAI) (1.5 T1 vs 1.0 T2; $Z = -2.22$, $p = .026$, $r = .17$), the difference in score was small (0.5). There was no significant association between age or gender and RInAB, DAI or SDQ score, therefore these were not considered confounders of stability.

Multimodal, multidisciplinary (Psychiatrist & Clinical Psychologist) diagnoses were made based on the above information. Based on clinical diagnoses, RAD was prevalent in 5.0% ($n=5$, 95% CI [0.7 – 9.3]) at T1 and 1.0% ($n=1$, 95% CI [below 0 – 2.9]) met criteria for a borderline diagnosis. At T2, 2.6% ($n=2$, 95% CI [below 0 – 5.2]) met criteria for a clinical diagnosis of RAD and 3.9% ($n=3$, 95% CI [below 0 – 8.3]) met criteria for a borderline diagnosis. It should be noted that in four cases, multi-disciplinary diagnoses of RAD were made in the absence of DAI criteria being met because observational data were so striking.

Observed (RInAB) and carer reported (DAI) RAD scores were investigated over time for test-retest reliability. RInAB scores at T1 and T2 were significantly associated ($\rho = .43$, $p < .001$, $r_s = .19$), test re-test reliability was found to be low. DAI scores at T1 and T2 were also found to be significantly associated ($\rho = .33$, $p = .005$, $r_s = .11$) and similarly, test re-test reliability was not supported.

Although the severity of RAD symptoms was low overall, results demonstrated that at T2 84% ($n = 63$) of participating children's observed RAD symptoms had not reliably changed, 8% ($n = 6$) had worsened (although mostly not to clinical levels) and 8% ($n = 6$) had improved compared to T1. In terms of carer reported scores at T2, 61% ($n = 42$) of participants showed no reliable change in their DAI score, 27% ($n = 19$) of children's symptoms had improved and 12% ($n = 8$) had worsened in comparison to T1. Reliable change was defined as a change in RInAB score of $> \pm 0.5$ between T1 and T2. In relation to carer-reported RAD symptoms, reliable change was defined as a change of $> \pm 1$ DAI standard score between T1 and T2.

An independent samples Mann-Whitney U test demonstrated no notable or statistically significant difference in observed or reported RAD symptoms between children who had returned to birth parents at T2 ($n = 7$) and children who had stayed in placements ($n = 69$), although numbers returning to birth parents were small (observed: *Median* = 0.48 (0.44, 0.66) versus *Median* = .44 (0.25, 0.67); $U(75) = 226.5$, $Z = -.771$, $p = .44$; $r = .01$) (carer reported: *Median* = .50 (0, 1.5) versus *Median* = 1.0 (0.0, 2.0); $U(72) = 247.0$, $Z = -1.142$, $p = .25$; $r = .02$).

Research Question 2: RAD symptoms and associated mental health and cognitive functioning

In order to explore demographic, mental health and cognitive functioning variables that may be associated with symptoms of RAD, correlations were carried out. Spearman's Rho correlations between all variables of interest are shown in Table 4. A significant association was found between observed RAD symptoms at T2 and Full Scale IQ ($\rho = -.28$, $n = 71$, $p = .017$) and Verbal IQ ($\rho = -.31$, $n = 71$, $p = .009$). Carer-reported RAD symptoms at T2 were significantly

associated with SDQ total difficulties ($\rho = .33$, $n = 50$, $p = .02$). No significant associations were found between T1 or T2 carer-reported RAD symptoms and placement moves ($p = .52$, $p = .65$).

Research Question 3: Concordance between observed and carer report of RAD symptoms

Five percent ($n = 5$, 95% CI [0.7-9.3]) of participants met observational criteria for RAD and 2.1% ($n = 2$, 95% CI [below 0 - 4.7]) met carer-report criteria for RAD at T1. Only 1.0% of children ($n = 1$, 95% CI [below 0 - 5.21]) met both observational and carer report diagnostic criteria for RAD at T1. At T2, 3.9% ($n = 3$, 95% CI [below 0- 8.3]) met observational criteria and 2.7% ($n = 2$, 95% CI [below 0 - 5.21]) met carer-report criteria for RAD (Figure 2 illustrates these findings). No children met both carer-reported diagnostic criteria and observational diagnostic criteria at T2; however for one of the children meeting observational criteria at T2, a carer-report measure of RAD was not available. See Table 3 for frequencies of participants who did and did not meet diagnostic criteria for RAD. According to Spearman's Rho correlations, no significant associations were found between observed RAD symptoms and carer-reported RAD symptoms at T1 ($\rho = .18$, $p = .081$) or T2 ($\rho = .19$, $p = .111$). The relationships between carer reported and observed symptoms were also investigated for when the observed symptoms were and were not informed by an SSP, non-significant associations remained (T1: $\rho = .39$, $p = .34$ versus $\rho = .17$; $p = .12$) (T2: $\rho = .18$, $p = .22$ versus $\rho = .20$, $p = .38$).

Discussion

The data of this explorative study supported a degree of stability in RAD symptoms and diagnosis, although a larger sample size along with diagnoses informed by fully validated measures, would allow for more certainty in addressing this. Based on clinical diagnoses, it was found that RAD 5.0% ($n = 5$) met diagnostic criteria for RAD and 1.0% ($n = 1$) met criteria for a borderline diagnosis at T1. At T2, 2.6% ($n = 2$) met diagnostic criteria and 3.9% ($n = 3$) met criteria

for a borderline diagnosis. This is in line with findings reported in Zimmerman (2016) and Gleason et al. (2011).

Only one child (1.0%) met both observational and carer-report diagnostic criteria for RAD at T1 and 0% ($n=0$) at T2. It is important to note however that at T2, three children continued to meet observational criteria for RAD and a further two children continued to meet carer-report criteria for RAD; despite all care placements being considered satisfactory (as assessed by the local authority). This may begin to address one of the most controversial aspects of the field, namely whether or not RAD is a state in response to current maltreatment or a disorder that persistent across contexts (including a nurturing care placement). The prevalence of RAD appeared to be very low, even in this high-risk cohort. Further, the confidence limits of the prevalence in the study were very large, suggesting a much larger sample would be needed to ascertain a reliable prevalence of RAD. It is possible that previous studies with relatively small sample sizes finding no persistent cases would have detected persistence with a larger sample size.

Overall, after around one year in an improved care setting, carer-reported RAD symptoms decreased significantly. This was not found to be the case for observed symptoms of RAD, where no significant difference was found between scores at T1 and T2. Results demonstrated that at T2 84% ($n= 63$) of participating children's observed RAD symptoms had not reliably changed, nor had 61% ($n= 42$) of participants according to carer report. It is important to note however that observed and carer reported symptom level was fairly low to begin with. Observed RAD symptoms at T1 and T2 were found to be significantly associated as were carer reported symptoms at T1 and T2. RInAB and DAI scores did not correlate with each other at T1 or T2, however.

Observed RAD symptoms at T2 were associated with Full Scale IQ and verbal IQ, Smyke et al. (2012) found similar findings and further research is necessary to understand the

links between verbal IQ and RAD, including the direction of causality. Carer-reported RAD symptoms at T2 were associated with more SDQ total difficulties ($r_{ho} = .34$).

Measurement of RAD is still at an early stage. Although parent-report RAD symptoms can be discriminated from other mental health presentations (Minnis, et al., 2007), the RInAB measure may have been picking up on symptoms that are not specific to RAD. An item referring to the child being uncomfortable with the situation, for instance could reflect anxiety within the context of appropriate attachment behaviours. The utilisation of a longitudinal study design comparing maltreated children with RAD to a typically developing group and to children with other disorders would be helpful in addressing this. The lack of significant associations between observed and carer-reported RAD symptoms at T1 or T2 is intriguing. According to carer report, symptoms of RAD were certainly less frequent than what was observed by trained raters. In order to explore this further, a much larger sample would be required whereby sensitivity of the RInAB and DAI would be determined against multimodal diagnosis using both measures; ideally also ratified by an experienced clinician. It is possible that at T1, carers did not know the child well enough to provide an informed rating, but this does not explain the discrepancy between observed and carer-reported symptoms at T2. It might be that there were biases affecting carers responses, for instance social desirability or eagerness to demonstrate a strengths based view of the child. It is also possible that foster carers lack awareness of RAD symptoms which are subtle in comparison to, for example, conduct problems. Parent/carer interaction has been described as characterised by “serve and return” where a sensitive carer responds to the “serve” of the child (<http://developingchild.harvard.edu/science/key-concepts/serve-and-return/>). If the child is not “serving”, as in RAD, the relationship between carer and child might fail to develop yet the carer may not notice the lack of any prompting to “return”. It may therefore be that, unlike most child psychiatric disorders, carer-report is problematic in RAD and observation may assume critical importance.

Limitations

The current study has a number of limitations. A few children had moved placement between T1 and T2 and a small number of children had returned back to birth parents. Although number of placements or returning to birth parents had no statistically significant impact, it is possible that a larger sample size may have illustrated a difference. Additionally, a notable number of participants had dropped out at T2. Although no significant differences were found between those who dropped out and those who remained in the study, selective attrition or factors that were not investigated may be playing a role and potentially affecting validity.

The study is further statistically limited due to the modest sample size, low prevalence of RAD symptoms across the sample and only singular cases of children meeting diagnostic criteria. For a rare disorder such as RAD, a much larger sample size or a different epidemiological technique (such as surveillance) would be required in order to be confident about prevalence rates and correlates.

In consideration of the assessment procedure of observed RAD symptoms, it was not always possible to be blinded to whether the video footage was T1 or T2, despite attempts. In addition, it could be a limitation that the procedure used to elicit attachment behaviours was not fully standardised. It was difficult at times to rate items relating to attachment due to children showing no apparent distress. Although participants were subject to procedures purposefully designed to elicit distress and activate attachment behaviours, noticeable distress was often still lacking. It could therefore be argued that the procedure was not stressful enough to elicit attachment behaviours. It could be considered a limitation that at T1, children had been with their carer for around three months. The DAI recommends carers know the child well and the RInAB authors recommend the child has been in placement for more than 6 months. In some cases, the child was moved to be with a different carer following T1, which could impact on attachment behaviours exhibited at T2 and make comparisons between T1 and T2 difficult. It is possible that

the validity of the observational rating was compromised as the children had not been with their new caregivers long enough to develop appropriate attachments. It is understandable that more intense negative emotions and atypical behaviors may have been demonstrated given the children would still be adjusting to their new setting. Nonetheless, one of the study aims was to explore change in RAD symptoms soon after placement in foster care; waiting longer (e.g. six months) may mean a crucial insight into the development or resolution of RAD symptoms in maltreated children coming in to care is missed.

The recently developed RInAB scale is the only known observational assessment of RAD. However it may be that the change in observed RAD scores over time is due to lack of test-retest reliability data for the scale rather than an actual change in level of RAD symptoms. In addition, the scale has not been normed on non-maltreated populations. It was noted that the rating of one item in the RInAB scale could be influenced by the caregivers reaction rather than the child's behaviour ('The child's behavior does not tend to elicit care and nurturing behavior from the caregiver'), perhaps the authors could consider slightly rewording the item.

Clinical Implications

If persistent, symptoms of RAD are likely to have profoundly negative effects on children's development as children who are emotionally withdrawn and inhibited are unlikely to elicit the kind of parental support needed for development (Prior & Glaser, 2006). Findings from the current study hopefully begin to provide a greater insight in to the occurrence and correlates of RAD, thus improving awareness of the disorder and any associated difficulties. It is important that professionals working with children, particularly those who may have been maltreated, are aware of the clinical symptoms and potential correlates of RAD; especially given emotional withdrawal is at the core of the disorder and such children are easily missed.

Future Directions

It is clear from the findings of this study that, it is difficult to draw firm conclusions about RAD and its correlates given its rarity. Larger samples and/or the pooling of samples

across studies are required in order to address important questions, as is further validation of the available observational measure. It would be helpful if future research could determine whether RAD predicts developmental problems over and above maltreatment itself. Furthermore, it would be useful to distinguish risk factors specific to RAD and therefore inform which children are at risk of persistent RAD. This would be an important step towards developing effective interventions for and ultimately preventing RAD.

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Table 1.

Demographics of sample

		Time 1 (n=100)	Time 2 (n=76)
Age (months)	mean (SD)	36 (14.7)	50 (14.3)
	min, max	12, 62	22, 77
Placement, n (%)	Foster carer no. 1	96 (96%)	56 (73.1%)
	Foster carer no. 2	2 (2%)	10 (13.2%)
	Adoptive family	2 (2%)	2 (2.6%)
	Kinship carer	0 (0%)	1 (1.3%)
	Birth parent	0 (0%)	7 (9.2%)
Gender, n (%)	Male	41 (41%)	27 (36%)
	Female	59 (59%)	49 (64%)

Table 2.

Descriptive Statistics of outcomes at T1 and T2. Variables are summarised as median (Q1, Q3) or mean (SD).

Measure	Time 1 (n=100)	Time 2 (n=76)
<i>Median (interquartile range) or mean (SD)</i>		
RInAB score	0.56 (0.31, 0.91)	0.46 (0.25, 0.67)
DAI score	1.50 (0.00, 3.25)	1.00 (0.00, 2.00)
Full-scale IQ	-	88.7 (12.5)
Verbal IQ	-	91.0 (11.1)
Performance IQ	-	89.9 (13.6)
SDQ total difficulty score	-	14.0 (11.0, 19.0)

Table 3

Categorical classifications of RAD symptoms

		Time 1		Time 2	
Meets observational cut off criteria (RInAB)	Yes	5	Median 2.41 (2.26, 3.69)	3	Median 2.01 (1.88, 2.38)
	No	95	Median 0.54 (0.29, 0.86)	73	Median 0.42 (0.25, 0.63)
Total		100		76	
Meets carer report cut off criteria (DAI)	Yes	2	Median 7 (6.5, 7.5)	2	Median 3.3 (2.25, 4.75)
	No	93	Median 1.5 (0.0, 3.3)	69	Median 1.0 (0.0, 2.0)
Total		95		71	
Clinical diagnosis Given	Yes				
	Borderline	5		2	
	No	1		3	
		0		0	
Total		5		2	

Table 4

Spearman's Rho Correlations between variables of interest

Measure		DAI score T1	DAI score T2	RInAB score T1	RInAB score T2
DAI score T1	Correlation Coefficient		.327**	.181	.064
	Sig. (2-tailed)	.	.005	.081	.585
	N		71	94	74
DAI score T2	Correlation Coefficient	.327**		.142	.192
	Sig. (2-tailed)	.005	.	.230	.111
	N	71		73	70
RInAB score T1	Correlation Coefficient	.181	.142		-.431**
	Sig. (2-tailed)	.081	.230	.	.000
	N	94	73		76
RInAB score T2	Correlation Coefficient	.064	.192	.431**	
	Sig. (2-tailed)	.585	.111	0.00	.
	N	74	70	76	
SDQ total difficultie s T2	Correlation Coefficient	.036	.329*	-.049	.078
	Sig. (2-tailed)	.805	.020	.732	.599
	N	52	50	52	48
WPPSI full scale IQ	Correlation Coefficient	-.108	-.162	-.085	-.283*
	Sig. (2-tailed)	.368	.188	.471	.017
	N	72	68	74	71
WPPSI verbal IQ	Correlation Coefficient	-.136	-.160	-.085	-.306**
	Sig. (2-tailed)	.256	.191	.473	.009
	N	72	68	74	71

**. Correlation is significant at the 0.01 level (2-tailed).

*. Correlation is significant at the 0.05 level (2-tailed).

Figure 1.

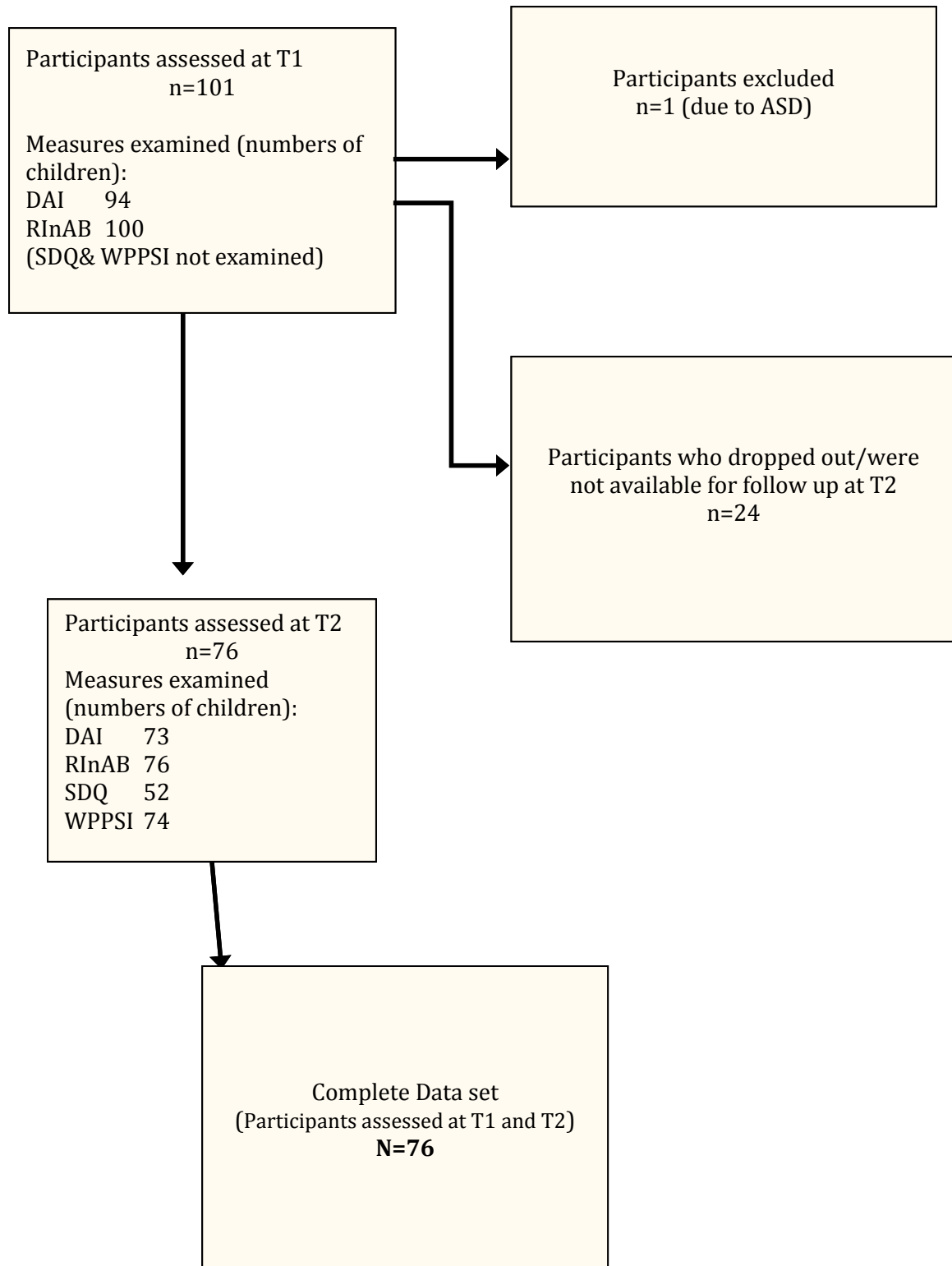
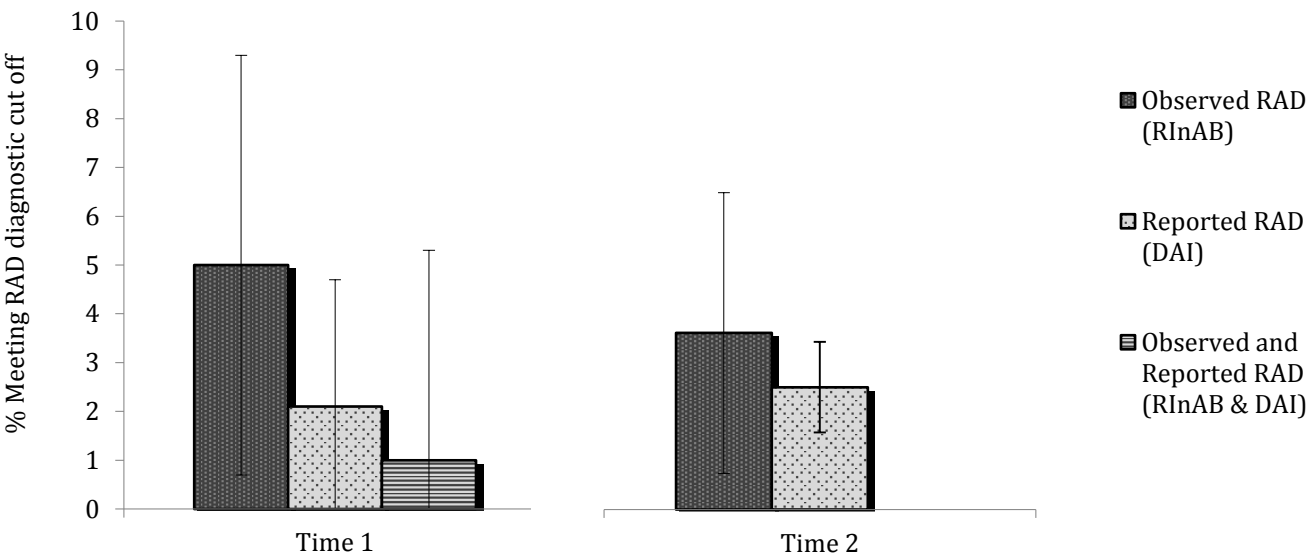
Flow chart of study participants

Figure 2.
Categorical diagnostic prevalence of RAD



APPENDICES

Appendix 1.1 RInAB observational tool



Rating of Inhibited Attachment Behavior – RInAB Version 4.0

ID _____

Strange Situation Procedure ☐

Caregiver-child interaction ☐

Other procedure _____

0 – Not at all characteristic of this child

2 – Somewhat characteristic of this child

4 – Very characteristic of this child

NA – Not applicable

	0	1	2	3	4	NA
A. Attachment Behavior						
1. When in distress, the child does not seek comfort from the caregiver						
2. When in distress, the child does not respond to comfort offered by the caregiver						
3. The child shows lack of a preference: no difference in the child's behavior with the stranger and the caregiver						
4. No evidence of arousal on caregiver's departure or reunion						
5. The child's behavior does not tend to elicit care and nurturing behavior from the caregiver						
B. Exploratory Behavior						
1. The child is uncomfortable with the situation						
2. The child is uncomfortable with the presence of the stranger						
3. The child spends more time in solitary play than in interactive play						
4. The child does not respond to the caregiver's initiatives for play.						
C. Socioemotional Behavior (Within the Relationship context)						
1. The child shows withdrawing behaviors						
2. The child shows a reduced or absent social and emotional reciprocity						
3. The child is passively compliant with others requests						
4. The child shows aggressive reactions or irritability						
5. The child shows an apparent misery, sadness, apathy and/or passivity						
6. The child is hypervigilant and/or fearful						
7. The child shows limited positive affect in moments where it would be expected						
8. The child does not initiate social interactions						